

Systematic Review

Clinical outcomes of lingual orthodontic treatment: a systematic review

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Summary

Objectives: To assess the available evidence on the effectiveness of lingual orthodontic treatment and related clinical parameters through a systematic review of relevant studies.

Materials and methods: Eligible clinical studies published from January 2000 to March 2015 were identified through electronic (five major databases) and hand searches. Risk of bias was assessed using the Cochrane risk of bias tool for prospective studies and a specially designed tool for retrospective studies.

Results: From the 3734 articles identified by the search, after application of specific inclusion and exclusion criteria, 16 papers were included in the study. Eleven studies were retrospective, four were prospective, and only one was a RCT. In detail, six studies evaluated differences of the treatment outcome from the pre-treatment set-up prediction, two studies evaluated the effect of treatment on periodontal and microbial parameters, and 10 studies assessed various clinical treatment related parameters. Despite several promising findings, the quality of evidence supporting them was found to be low in most cases.

Conclusions: This systematic review showed encouraging results on the clinical outcome of lingual orthodontic treatment, especially in regards to the achievement of individualized treatment goals and the reduction of decalcifications on the bonded surfaces of the teeth. However, additional well-designed prospective clinical trials with larger samples are needed to confirm those findings. Several aspects of lingual orthodontic treatment were difficult to be conclusively evaluated due to the study design, the heterogeneity, the small samples sizes, and the high risk of bias seen in the majority of the included studies.

Introduction

The aesthetic demands of patients have been increased during the years regarding not only the treatment objectives, but also the influence of orthodontic appliances in patient's aesthetic appearance. Conventional orthodontic treatment has been shown to compromise facial appearance (1) and this is a major concern of patients seeking orthodontic treatment (2). As a result, aesthetic materials and techniques have been introduced in clinical practice to overcome these limitations (3).

Lingual orthodontics comprise a fundamental expression of this necessity (4). Since its introduction in 1980s, several systems and techniques have been introduced, revealing the increased interest of patients and doctors for this treatment approach (5). The major advantage of lingual orthodontics is that the appliances are not visible. However, lingual orthodontics comprises a relatively new treatment option that has certain differences to the well-established labial orthodontic treatment, mainly attributed to the position of the appliances (6, 7). A recent systematic review of six studies that compared certain adverse effects of

lingual and labial orthodontic treatment (8) provided further support to this viewpoint. Differences that may exist in various aspects of treatment, and can be related to the patient, the practitioner, or the appliance itself, might be responsible for the limited, although increasing use of lingual orthodontics in everyday practice (5, 7, 9, 10).

During the years, several case reports, reviews, and clinical research papers have discussed various aspects of lingual orthodontic treatment. However, a thorough systematic evaluation of clinical studies related to clinical outcomes of lingual orthodontic treatment is lacking from the literature. Such a review will allow gather the supporting evidence on this expanding treatment approach, will help clinicians to provide evidence-based treatment, and will provide recommendations for targeted future research in the field. Thus, the purpose of the present study is to assess the available evidence on the effectiveness of lingual orthodontic treatment and related clinical parameters through a systematic review of recent relevant studies (published later than 1 January 2000).

Material and methods

The present systematic review was conducted and reported according to the Preferred Reporting Items for Systematic reviews and Meta-Analyses (PRISMA) guidelines for reporting studies that evaluate health care interventions (11, 12), to the Meta-analysis of Observational Studies in Epidemiology (MOOSE) guidelines for reporting observational studies (13, 14), and to the Cochrane Non-Randomized Studies Methods Group (15). The Grades of Recommendation, Assessment, Development, and Evaluation (GRADE) approach (16) was used for grading the quality of evidence. A pilot Pubmed search followed by systematic evaluation of 10 potentially eligible and randomly selected studies were performed in order to draft the review protocol. Methods of search and analysis, eligibility assessment, data extraction forms, and all other methods and tools used were defined in advance of the study based on the results of the pilot study.

The main type of intervention, e.g. buccal or lingual-fixed orthodontic appliances is unlikely to be randomized, since patients usually decide about this issue based on individual needs and preferences. Furthermore, this is a relatively new and unexplored treatment, especially in terms of systematic evaluations. Thus, we decided to evaluate for eligibility also non-randomized and retrospective studies following a quality assessment protocol (see details below).

Search methods for identification of studies

Electronic search was conducted by two authors (G.V. and N.G.) in five major databases, Medline, EMBASE, Google scholar beta, all Cochrane Databases, and Conference Paper Index with the last update on March 2015. A complex Medline search (via Pubmed) was performed to identify any relevant study, based upon various combinations of key words including: 'lingual orthodontics', 'lingual patients', 'lingual brackets', in all fields (Limits: Humans, English, German, French, Italian). A detailed description of the Medline electronic search strategy is provided in [supplemental Appendix 1](#). Similar searches were performed in all the electronic databases used for the study.

All papers assessed through full text reading, all included papers, and all reviews identified were subjected to hand search of their references for relevant papers that might have been missed during the electronic search.

Unpublished literature was searched through Cochrane Central Register of Controlled Trials, Conference Paper Index, and Google scholar.

Selection of studies

Eligibility assessment was performed in a standardized manner, independently, and in duplicate by two reviewers (H.K. and G.V.) who were not blinded to the identity of the authors, their institution, or the results of the research. Any disagreement was resolved by consensus and through discussion among all authors. Titles and abstracts were screened first and afterwards full text review of any relevant and potential for inclusion article was conducted.

A positive exclusion method was used, whereby only those publications that did not meet one or more of the inclusion criteria were excluded. An independent reviewer (I.M.) checked a random selection (20 per cent) of filtered articles for consistency. Inter-rater agreement on study eligibility was assessed by Cohen's kappa.

Eligibility criteria

The following inclusion criteria were applied:

1. Randomized clinical trials (RCTs), prospective controlled clinical trials (CCTs), or retrospective studies with a control group reporting on results or treatment parameters related to orthodontic treatment performed with fixed lingual brackets bonded at least to all teeth between first molars, in one or both arches.
2. English, German, French, and Italian languages.

The exclusion criteria were:

1. Studies reporting results emerging from questionnaires.
2. *In vitro* and animal studies.
3. Case reports/case series (sample size ≤ 10).
4. Editorials, opinions, reviews, and technique description articles, without reported sample.
5. Studies referring to lingual treatment performed in selected teeth of an arch or combined with other appliances, such as labial brackets, in a non-defined manner and not in order to investigate a specific hypothesis under a split-mouth design.
6. Studies published prior to January 2000 were excluded to control for confounding derived from the large differences of bracket types and application techniques of that time compared with the current practice.

Types of participants

Healthy participants who require orthodontic treatment with fixed appliances with no age limit. Studies including patients receiving any kind of medication, which can influence orthodontic treatment or patients receiving orthognathic surgery, syndromic patients, patients with cleft lip and palate, or any systemic disease were excluded.

Types of interventions

Any treatment approach utilizing orthodontic fixed lingual appliances placed in one or both dental arches, used to correct any type of malocclusion, were included.

Control

Conventional orthodontic treatment with fixed labial appliances or no treatment. The planned treatment outcome and comparable patients receiving fixed appliance treatment with different lingual bracket systems or application techniques (i.e. different bonding protocols) were also considered.

Types of outcome measures

Any measure of performance or effectiveness of the intervention tested including its accuracy and any parameter regarding the quality

of the treatment outcome, periodontal, or dental parameters (plaque index, gingival index, root resorption, tooth necrosis/loss of vitality etc.), or various clinical aspects of treatment (duration of treatment, stability of final outcome, bond failures etc.).

Data extraction process

Data extraction was performed by two authors (I.M. and H.K.), independently in the pre-determined data extraction forms that were also used for quality assessment of the included studies. In cases of inconsistencies re-examination of the original studies by the two reviewers and discussion resolved any disagreements. N.G. was responsible for checking the data extraction forms. Inter-rater agreement on data extraction was assessed by Cohen's kappa.

Quality assessment of individual studies

The quality assessment of the eligible studies was performed by two investigators, independently (D.K. and N.G.). In areas of disagreements, a joint decision was obtained after discussion between all authors in order to reach a consensus. Quality assessment of randomized studies was performed using the Cochrane Risk of bias tool (17). The same tool was also used for non-randomized prospective studies in the applicable domains.

To assess the quality of the retrospective studies, two investigators working independently (D.K. and N.G.) evaluated several items according to a modified quality assessment tool designed based on Newcastle–Ottawa scale (18), relevant guidelines of the *Cochrane Handbook for Systematic Reviews of Interventions* Version 5.1.0 (15), and the GRADE approach (16).

We categorized overall risk of bias in any included study according to the following.

1. Low risk of bias (plausible bias unlikely to seriously alter the results) if all key domains were assessed as at low risk of bias.
2. Unclear risk of bias (plausible bias that raises some doubt about the results) if one or more key domains were assessed as at unclear risk of bias.
3. High risk of bias (plausible bias that seriously weakens confidence in the results) if one or more key domains were assessed as at high risk of bias.

Dealing with missing data

The analysis was performed using only the available data (ignoring missing data); methods for estimating missing SDs as described in Section 7.7.3 of the *Cochrane Handbook for Systematic Reviews of Interventions* (15), were used where applicable.

Data synthesis

A meta-analysis was planned only if there were at least two studies of similar comparisons, reporting similar outcome at similar time points. For qualitative synthesis, the included studies were to be assigned to general groups according to their subject category.

Determination of available evidence supporting clinical recommendations

Following the quality assessment of individual articles, each one was assigned to a group according to the studied subject. For each subject, the overall quality of the body of evidence was determined after considering the quality (assessment of individual studies), quantity (magnitude of treatment effect, number of studies, sample size across studies), and consistency (the extent of similarity of different studies

in their findings) of the available studies and their findings on the subject. Clinical recommendations were formulated based on these considerations and by balancing the desirable and undesirable consequences of each intervention.

Results

Literature flow

The initial electronic search yielded 3734 studies. After reading titles, abstracts, and application of inclusion and exclusion criteria, 87 studies were examined in their full-text forms. From these, 16 studies were deemed eligible for inclusion in the review and for qualitative and/or quantitative synthesis (6, 18–32). The flow chart of study selection together with reasons for exclusion is provided in Figure 1. The kappa scores for the initial selection and data extraction procedures were 0.86 and 0.91, respectively.

Characteristics of included studies

The 16 included studies were assigned to three general groups according to the main outcome tested: 1. accuracy of treatment prediction, 2. periodontal parameters, and 3. other clinical parameters. Two included studies were assigned in two groups.

Eleven studies were retrospective, four were prospective CCTs and one was RCT. Six retrospective studies tested the accuracy of lingual treatment through the comparison of set-ups with actual treatment outcome (19–23, 29). One of them also tested the occlusal outcome of lingual treatment compared with labial treatment (23). Ten studies tested various clinical aspects of treatment, such as effectiveness, bond failures, formation of white spot lesions, anchorage loss, and treatment duration (6, 18, 22–25, 28, 30–32). Finally, two prospective CCTs assessed treatment effects on various periodontal parameters, such as the plaque index and the gingival index (26, 27). An overview of the characteristics of the included studies is provided in Table 1.

Publication bias

Statistical analysis of publication bias was not indicated, since no quantitative synthesis was undertaken.

Risk of bias of included studies

For non-randomized prospective CCT studies, the items of random sequence generation and allocation concealment were not applicable and were set by default as unclear. This decision was based on the specific characteristics of the clinical question studied; primarily the inability of personnel to assign patients in groups and/or predict favourable versus unfavourable outcomes. Initial inter-rater disagreement existed in 3 out of 16 cases (agreement greater than 80 per cent) and these were all between unclear and high risk ratings. Disagreements were resolved through discussion by all authors until consensus was reached.

RCTs

Only one RCT was included and was assessed as unclear overall risk of bias (6). Figure 2 shows the summary of risk of bias assessment for the RCT study according to the Cochrane Risk of bias tool.

Prospective CCTs

From the four CCTs included, two were assessed as unclear (26, 27) and two as high overall risk of bias (25, 30). Figure 2 shows the summary of risk of bias assessment for non-randomized prospective CCTs.

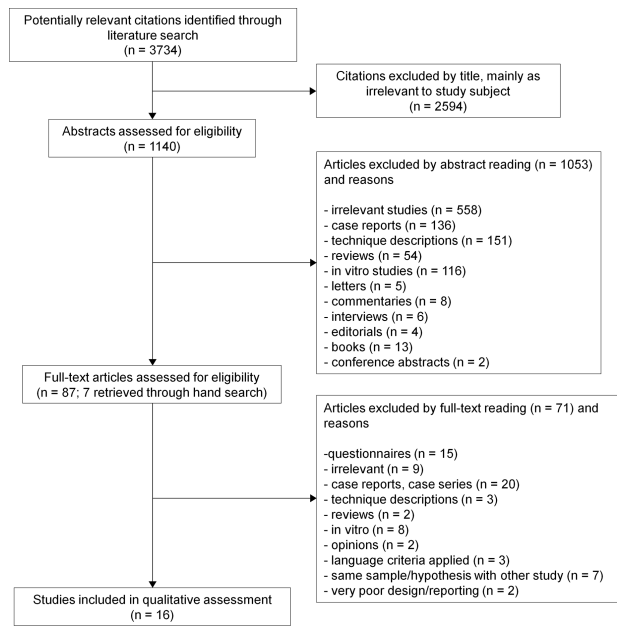


Figure 1. Flow chart of study selection.

Retrospective studies

From the 11 retrospective studies included, 10 were assessed as high (18, 20–24, 28, 29, 31, 32) and 1 as unclear overall risk of bias (19). Table 3 shows the assessment of risk of bias for retrospective studies.

Qualitative synthesis of included studies

A meta-analysis was not feasible since there were no studies of similar comparisons, reporting similar outcomes at similar time points. In order to proceed to the qualitative analysis, the 16 studies were divided into three main groups according to the aspect of lingual treatment tested. The aforementioned categorization did not derive from any established methodological background, but was implemented based on clinical interest. Furthermore, there is a variety of outcome measures reported in each study that does not allow in most cases for standardized reporting of results. An overview of the set-up, the findings, and the overall risk of bias assessment of individual studies is provided in Tables 1 and 2.

Effects of interventions

Accuracy testing

In total, six studies tested the accuracy of lingual orthodontic treatment. All utilized a retrospective study design and tested the Incognito System (3M Unitek, Monrovia, California, USA) (19–23, 29). Five of them had high (20–23, 29) and one unclear risk of bias (19) (Table 3). Two studies (19, 20) assessed the deviations in tooth position between the planned (setup) and the actual treatment outcome measured on three-dimensional (3D) digital dental casts. Both agreed that in most cases anterior teeth were positioned within 1 mm and within 4 degrees of their planned positions, whereas differences increased from anterior to posterior, with the second molars showing the poorest results. Another study tested the deviations in lower intercanine distance between the predicted (setup) and the actual outcome measured on 3D digital dental casts and reported a difference of 0.44 (SD: 0.29) mm (21). A study that tested the deviations in lower incisor inclination between the planned (setup) and the actual outcome in Class III patients reported a difference of 3.75 degrees (SD: 3.06 degrees) (29). This difference was increasing in extraction cases and when undersized finishing wires were placed.

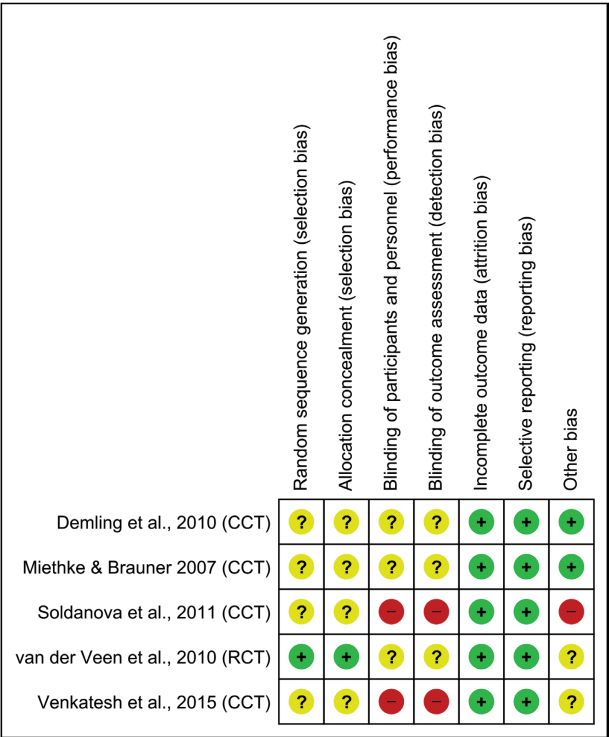


Figure 2. Risk of bias summary for included RCT and prospective CCT studies. The plus sign indicates low risk of bias; the circle with question mark indicates unclear risk of bias; the minus sign indicates high risk of bias. Overall, studies with at least one high are considered high risk of bias, studies with at least one question mark unclear risk of bias, while studies with plus signs only low risk of bias. For CCTs, the first two items are not applicable (default: unclear).

Central incisor apices also tended to move at the lingual side of the symphysis during incisor decompensation. One study tested the accuracy of the Incognito lingual technique + Herbst on mandibular incisor proclination and reported a difference between planned and final incisor positions of 2.2 degrees (SD: 1.0 degrees) ($P > 0.05$) (22). Finally, one study tested the deviations in peer assessment rating (PAR) scores measured on dental casts between the setup and the actual lingual treatment outcome and reported a difference of 4.1 in total weighted PAR score ($P < 0.05$) (23).

Clinical parameters testing

In total, 10 studies tested various clinical aspects of treatment, such as effectiveness, bond failures, formation of white spot lesions, anchorage loss, or treatment duration. Nine of them had high (18, 22–25, 28, 30–32) and one unclear risk of bias (6) (Table 3; Figure 2). A retrospective study on the effect of the Incognito lingual technique + Herbst reported a change in mandibular incisor inclination of -16.5 degrees to 5.8 degrees that was according the planned movement in each case (22). Another retrospective study compared the occlusal outcome and treatment duration with lingual (Incognito) and labial appliances and did not report any significant difference ($P > 0.05$), although there was a tendency for higher improvement in weighted PAR index through labial treatment (23). One retrospective study tested potential differences in anchorage loss in premolar extraction cases between lingual bidimensional preadjusted brackets (Ormco Corp, Glendora, California, USA) and labial appliances and reported almost double amount of anchorage loss in labial treatment (24). The same issue

was tested in a prospective CCT study that used another bracket system (STb, Ormco, Orange, California, USA) and yielded similar results (30). A prospective study tested changes in the lower dental arch of Class I patients with crowding following orthodontic therapy with labial or lingual two-dimensional (2D) technique (Forestadent, St Louis, Missouri, USA). The only significant difference regarded the position of incisors relative to A–Po (lingual: 1.0, SD: 0.5 mm; labial: 0.0, SD: 1.5 mm, $P < 0.05$) (25). The only included RCT study tested the development of white spot lesions during lingual (Incognito) and labial appliance treatment. This study utilized a split mouth design and was characterized as unclear risk of bias mainly because blinding was not possible and this might have resulted to performance bias. The study concluded that the number of buccal caries that formed or progressed during treatment was 4.8 times higher than the number of lingual lesions ($P = 0.01$). Furthermore, there was an overall increase in caries extent/total lesion area that was more for buccal surfaces in a subject than for lingual surfaces ($P < 0.05$) (6). Another retrospective study from the same research group tested the effect of the addition of an extra layer of a hydrophilic resin, during bonding in the incidence of demineralization beneath the base of lingual brackets (Incognito). The addition of hydrophilic resin induced a 3-fold decrease in demineralization incidence after treatment (28). A retrospective study compared treatment with lingual (STb, Ormco) and labial appliances and found similar PAR reduction, treatment duration and root resorption with both techniques. The only difference was detected on post-treatment interincisal angle which was increased more by lingual treatment (T0–T1, labial: 11.2 degrees, SD: 11.6 degrees; lingual, 17.9 degrees, SD: 8.9 degrees, $P < 0.05$) (31). In another retrospective study, the number of lost brackets during the first year of treatment was found to be similar in lingual (Incognito) and labial treatment (32). Finally, a recent retrospective study showed that treatment duration was significantly smaller with one lingual bracket system (WIN; S1: 17.96 m, S2: 20.49 m) compared to another (Incognito; S1: 22.7 m, S2: 29.79 m) (18).

Periodontal parameters testing

Two prospective CCTs with unclear risk of bias assessed treatment effects on various periodontal parameters (26, 27). One split-mouth study tested clinical periodontal and microbial indices before and 4 weeks after bonding of Incognito lingual appliances. Plaque index and bleeding on probing significantly increased in this period in the bonded sites, while no difference was detected for probing depth. Periodontopathogenic bacteria did not change significantly (26). The other study assessed clinical periodontal parameters after at least 6 months on treatment, at three consecutive appointments (3–4 week interval), in patients treated with Ormco seventh generation lingual (Ormco Europe, Amersfoort, The Netherlands) or Invisalign appliances. All the indices apart from probing depth improved significantly between the first and third assessments in the Invisalign patients, while they were significantly worse in lingual treatment in all time points (27).

Discussion

The growing interest of the scientific community for lingual orthodontics over the years is evident by the increasing number of relevant publications in the literature. The search of the present systematic review covered a period of 15 years (2000–2015) and 13 out of the 16 included studies were published in the last 5 years. This underlines the need for orthodontics to meet the increased aesthetic demands expressed by patients (33). However, lingual orthodontic treatment

is still not considered a conventional option with widespread use in current orthodontic practice. Possible explanations for this could be the relatively unexplored clinical performance of the technique, the additional knowledge required to be applied, the fact that lingual orthodontics are not taught in the majority of postgraduate programs at the same extent as conventional buccal techniques, as well as the higher costs that are usually involved.

We decided to include only recent studies (from January 2000 and on) by applying time restriction to the search strategy in an attempt to be compatible with current clinical conditions. Otherwise, if older studies were included, the evolution of lingual appliances and techniques over the years (34, 35) could give results not applicable to the present clinical practice. The earliest included study was published in 2003 and the remaining studies were published after 2007.

The 16 included comparative studies were divided in three major groups according to the study subject. These regarded 1. accuracy, 2. clinical parameters, and 3. periodontal parameters testing.

In the first group, six retrospective studies tested the accuracy of lingual orthodontic treatment by comparing the planned treatment goal in the setup with the actual outcome obtained after treatment (19–23, 29). The results of these studies were encouraging, indicating that modern lingual orthodontic systems can achieve to a significant extent the individualized treatment goals established by the setup. The retrospective nature of all these studies, however, does not allow for elucidating the parameters that influence the achievement of the planned treatment outcome for the individual patient. For this, well-designed prospective clinical trials are necessary.

The second subject group included 10 studies that investigated various clinical aspects of lingual orthodontic treatment. The only available RCT showed that the number of buccal caries lesions that formed or progressed during buccal fixed appliance treatment was approximately five times higher than the number of lingual caries lesions in lingual treatment (6). However, since blinding was not possible because of the split-mouth study design, performance bias during the study period cannot be excluded. Because of the high importance of the results of this study for the dental health, additional RCTs with large samples are needed to confirm this positive effect of lingual appliance treatment. In another retrospective study, it was found that the incidence of demineralization under the lingual bracket base was considerably reduced in children and adolescents through the addition of hydrophilic resin during the bonding procedure (28).

The other studies in the second subject group investigated various issues including effectiveness, treatment duration, anchorage loss, and bond failures. A retrospective study tested in a small group of patients the effect of lingual technique + Herbst on mandibular incisor proclination and concluded that the incisors are positioned according to the planned movement (22). Another retrospective study reported similar occlusal outcome and treatment duration with lingual (Incognito) and labial appliances (23). Two studies tested differences in anchorage loss in premolar extraction cases between lingual (STb or bidimensional, Ormco) and labial appliances and reported almost double amount of anchorage loss in labial treatment (24, 30). One prospective study found that labial and lingual (Forestadent) 2D treatment induces in most cases similar changes to the lower dental arch in Class I patients with crowding (25). A retrospective study concluded that treatment with lingual (STb) and labial appliances result in similar PAR reduction, treatment duration, and root resorption (31). In another study, the number of lost brackets during the first year of treatment was found to be similar for lingual and labial appliances (32). Finally, a recent retrospective study compared two different lingual systems and showed that treatment

Table 1. Characteristics of included studies grouped according to their main subject: (a) accuracy testing, (b) various clinical aspects testing, and (c) periodontal parameters testing.

Study	Subject group	Main objective	Place of sample collection	Study design	Treatments tested and sample size	Method of sample recruitment	Main type of collected data
Grauer and Proffit, Am J Orthod Dentofacial Orthop 2011 (19)	Accuracy	Accuracy of Incognito lingual orthodontic technique	Bad Essen, Germany	Retrospective case series	Incognito lingual treatment ($n = 94$)	Consecutive patients debonded between January 2008 and January 2009	Tooth position in the setup and actual outcome measured at scanned dental casts
Pauls AH, J Orofac Orthop 2010 (20)	Accuracy	Accuracy of Incognito lingual orthodontic technique	Bad Essen, Germany	Retrospective case series	Incognito lingual treatment ($n = 25$)	Selection based on specific criteria	As above
Thalheim and Swestka-Polly, Inf Orthod Kieferorthop 2008 (21)	Accuracy	Accuracy of Incognito lingual orthodontic technique	Bad Essen, Germany	Retrospective case series	Incognito lingual treatment ($n = 20$)	Selection based on specific criteria	Lower intercanine distance in the setup and actual outcome measured at scanned dental casts
Lossdörfer <i>et al.</i> , J Orofac Orthop 2013 (29)	Accuracy	Accuracy of Incognito lingual orthodontic technique	Bad Essen, Germany	Retrospective case series	Incognito lingual treatment ($n = 34$)	Selection based on specific criteria	Lower incisor inclination in the setup and actual outcome measured at scanned dental casts superimposed into cephalograms
Wiechmann <i>et al.</i> , Head Face Med 2010 (22)	Accuracy and clinical	Effect of the Incognito lingual technique + Herbst on mandibular incisor proclination	Bad Essen, Germany	Retrospective case series	lingual treatment + Herbst ($n = 12$)	Selection based on specific criteria	Lower incisor position before treatment, in the setup and after treatment, measured at scanned dental casts
Smith C, dissertation 2010 (23)	Accuracy and clinical	Accuracy of Incognito lingual orthodontic technique; occlusal outcome and treatment duration with lingual and labial appliances	Illinois, USA	Retrospective case series	Incognito lingual ($n = 21$) versus various labial ($n = 22$)	Consecutive selection based on specific criteria	PAR scores measured in the setup and actual outcome (final) casts for lingual treatment and between initial and final casts for both groups
Geron <i>et al.</i> , Angle Orthod 2003 (24)	Clinical	Anchorage loss relative to extraction site (first versus second premolar), appliance (lingual versus labial), age (adolescents versus adults), overjet and crowding	Tel Aviv, Israel	Retrospective case series	Bidimensional lingual (Ormco; $n = 13$) versus labial (Victory System, 3M Unitek; $n = 15$)	Selection based on specific criteria	Anchorage loss (amount of mesial movement of the upper first molar) measured in cephalograms and dental casts
Venkatesh <i>et al.</i> , APOS Trends Orthod 2015 (30)	Clinical	Anchorage loss during first premolar extraction space closure with labial and lingual appliances	Karnataka, India	Prospective CCT	STb lingual (Ormco; $n = 10$) versus labial (Victory System, 3M Unitek; $n = 10$)	Selection based on specific criteria	Anchorage loss (amount of mesial movement of the upper first molar) measured in cephalograms

Table 1. Continued

Study	Subject group	Main objective	Place of sample collection	Study design	Treatments tested and sample size	Method of sample recruitment	Main type of collected data
Soldanova <i>et al.</i> , Eur J Orthod 2011 (25)	Clinical	Changes in the lower dental arch with the labial or the lingual 2D technique	Prague, Czech republic	Prospective CCT	lingual 2D (Forestadent; $n = 25$) versus labial (Minitrim Roth, Dentaurum; $n = 25$)	Selection based on specific criteria	Cephalometric measurements and images of dental casts obtained pre and post-treatment
van der Veen <i>et al.</i> , Eur J Oral Sci 2010 (6)	Clinical	Caries incidence after treatment with lingual or labial appliances	Bad Essen, Germany	RCT (split mouth)	Incognito lingual ($n = 14$) versus labial (Orthos, Ormco; $n = 14$)	Consecutive selection based on specific criteria	White spot caries lesions measured at white light photographs and quantitative light-induced fluorescence images pre and post-treatment
Beyling <i>et al.</i> , Head Face Med 2013 (28)	Clinical	To test the effect of the addition of an extra layer of a hydrophilic resin, during bonding, in the demineralization beneath the lingual bracket base	Bad Essen, Germany	Retrospective case series	Incognito lingual bonded conventionally ($n = 20$) versus bonded with an extra resin layer ($n = 20$)	Consecutive selection based on specific criteria	Sub bracket lesions measured at standardized photographs, pre and post-treatment, in the six maxillary anterior teeth
Deguchi <i>et al.</i> , Angle Orthod 2015 (31)	Clinical	Differences in cephalometric values, root resorption, occlusal indices, and functional aspects, between labial and lingual treatment	Japan	Retrospective case series	STb lingual (Ormco) ($n = 24$) versus labial ($n = 25$)	Consecutive selection based on specific criteria	Cephalometric values, degree of root resorption, PAR, objective grading system (OGS), and functional evaluation pre and post-treatment
Ziebur <i>et al.</i> , Am J Orthod Dentofacial Orthop 2014 (32)	Clinical	To compare frequency and position of bond failures in patients treated with lingual or buccal appliances	Munster, Germany	Retrospective case series	Incognito lingual ($n = 59$) versus Mini Diamond Brackets/Accent Molar Tubes (Ormco) labial ($n = 44$)	Selection based on specific criteria	Number and position of lost brackets in the first year of treatment
Knösel <i>et al.</i> , Head Face Med 2014 (18)	Clinical	To compare treatment duration with two types of customized lingual appliances	Bad Essen, Germany	Retrospective case series	Incognito lingual (3M Unitek) ($n = 220$) versus WIN lingual (DW LingualSystems) ($n = 156$)	Selection based on specific criteria	Treatment duration
Demling <i>et al.</i> , Angle Orthod 2010 (26)	Periodontal health	Short-term effect of lingual appliances on periodontal and microbial parameters	Hannover, Germany	Prospective CCT (control: maxilla and buccal sites)	Incognito lingual versus no appliances ($n = 20$; split mouth)	Consecutive selection based on specific criteria	Clinical periodontal evaluation and microbial analysis before and 4 weeks after bonding

Table 1. Continued

Study	Subject group	Main objective	Place of sample collection	Study design	Treatments tested and sample size	Method of sample recruitment	Main type of collected data
Miethke and Brauner, J Orofac Orthop 2007 (27)	Periodontal health	Periodontal health of patients treated with Invisalign or lingual appliances	Berlin, Germany	Prospective CCT (control: second, fourth quadrant)	Ormco 7gen lingual ($n = 30$) versus Invisalign ($n = 30$)	Consecutive selection based on specific criteria	Clinical periodontal evaluation after at least 6 months during treatment at three consecutive appointments (3–4 weeks interval)

Table 2. Results of individual studies, quality assessment, and synthesis of results.

Study	Subject group	Definition of pre-specified main outcome	Summary outcome data	Additional outcomes	Quality assessment*
Grauer and Profit, Am J Orthod Dentofacial Orthop 2011 (19)	Accuracy	Level of accuracy and effect of age, gender, crowding, overbite, overjet, ANB, days in treatment, days in slot-filling wire, Class II elastics, vertical elastics, interproximal reduction, rebondings, jaw, and tooth type	In most cases, anterior teeth were positioned within 1 mm and within 4° of their planned positions; differences increased from anterior to posterior; highly significant differences in all discrepancies except tooth long-axial rotation for the maxilla versus the mandible, and in all parameters for tooth type	Various parameters were statistically related to the amount of rotational and translational discrepancy, but each of these factors explained only a small amount of the total discrepancy	Unclear risk
Pauls AH, J Orofac Orthop 2010 (20)	Accuracy	Level of accuracy and effect of age, jaw, various finishing wires, bracket generation, original front tooth axial position, change in the front tooth inclination during lingual treatment, type of tooth, type of scanner	Mean deviations of set-up from end result were: tip (UJ/upper jaw 5.53°; LJ/lower jaw 5.72°), torque (UJ 5.16°; LJ 5.23°), supra/infraposition (UJ 0.89 mm; LJ 0.97 mm), rotation (UJ 5.82°; LJ 5.40°), in/out (UJ 1.19 mm; LJ 1.07 mm) and anterior/posterior position (UJ 1.04 mm; LJ 1.00 mm); significant difference between jaws in the antero-posterior position only	Significant differences for final archwires and different bracket generations and tooth types; the upper central incisors yielded the lowest values after the lower front teeth with deviations in rotation less than 4.5° and in translation less than 0.40 mm; from anterior to posterior, feasibility decreased by approx. 2° and 1 mm, with the second molars showing the poorest values	High risk
Thalheim and Swestka-Polly, Inf Orthod Kieferorthop 2008 (21)	Accuracy	Effect of lingual treatment on lower intercanine distance	Difference in intercanine distance of set-up versus final result 0.44 ± 0.29 mm	Difference in intercanine distance of initial versus final result 1.85 ± 1.58 mm	High risk
Lossdörfer <i>et al.</i> , J Orofac Orthop 2013 (29)	Accuracy	Changes in lower incisor inclination during treatment and differences between set-up and treatment result	Difference of clinically achieved versus planned inclinations was $5.89 \pm 5.43^\circ$, after correcting for changes in occlusal plane inclination. It was $3.75 \pm 3.06^\circ$ versus $8.59 \pm 6.58^\circ$ for full-size versus undersized finishing archwires and $7.88 \pm 6.36^\circ$ versus $4.65 \pm 4.48^\circ$ for extraction versus non-extraction cases	21 central incisor apices were located in the middle symphyseal third at baseline. Only 5 remained at debonding, whereas the other 16 moved to the lingual third ($P < 0.001$). A total of 13 apices were located in the lingual third at baseline and retained this position in all but one patient	High risk

Table 2. Continued

Study	Subject group	Definition of pre-specified main outcome	Summary outcome data	Additional outcomes	Quality assessment*
Wiechmann <i>et al.</i> , Head Face Med 2010 (22)	Accuracy and clinical	Changes in lower incisor inclination during lingual + Herbst treatment and differences between set-up and treatment result	Planned incisor inclination: -16.5° to 5.8° ; difference between planned and final incisor rotations was $2.2^\circ \pm 1.0^\circ$ ($P > 0.05$)	-	High risk
Smith C, dissertation 2010 (23)	Accuracy and clinical	PAR score in lingual and labial treatment	treatment time: lingual 22.35 ± 5.19 m, labial 22.35 ± 5.15 m (ns); total reduction in PAR score: lingual 15.24 ± 7.71 , labial 18.27 ± 11.15 (ns); percentage of PAR reduction: lingual $77.51 \pm 23.11\%$, labial $79.83 \pm 19.34\%$ (ns); diagnostic set-up (0.4) versus post-tx (4.5) total weighted PAR ($P < 0.05$)	There was no significant difference between any of the individual PAR components and in total weighted PAR scores in the two groups pre-treatment; the only difference post-treatment was in overbite ($P < 0.05$; labial mean PAR = 0; lingual = 0.29)	High risk
Geron <i>et al.</i> , Angle Orthod 2003 (24)	Clinical	Anchorage loss (amount of mesial movement of the upper first molar) during lingual treatment	Cephalograms: lingual = 1.84 ± 1.4 mm, labial = 3.0 ± 1.4 mm ($P < 0.05$); casts: lingual = 2.4 ± 1.9 , labial = 3.9 ± 2.7 (ns)	—	High risk
Venkatesh <i>et al.</i> , APOS Trends Orthod 2015 (30)	Clinical	Anchorage loss in first premolar extraction cases (mesial movement of the upper first molar)	Lingual: 1.24 ± 0.17 mm, labial: 2.06 ± 0.39 mm ($P = 0.001$)	—	High risk
Soldanova <i>et al.</i> , Eur J Orthod 2011 (25)	Clinical	Cephalometric and cast measurements in lingual and labial treatment	Significant difference in the position of incisors relative to A–Po (lingual: 1.0 ± 0.5 mm; labial: 0.0 ± 1.5 mm $P < 0.05$); no other differences in position of incisors, canines or molars	Active treatment time: lingual: 14.6 ± 6.98 m; labial: 15.44 ± 7.38 m	High risk
van der Veen <i>et al.</i> , Eur J Oral Sci 2010 (6)	Clinical	Presence/absence or progress of lesions in lingual and labial treatment	The number of caries that formed (buccal: $n = 16$, lingual: $n = 1$) or progressed (buccal: $n = 21$, lingual: $n = 7$) was higher in buccal compared to lingual lesions ($P = 0.01$); the overall increase in caries extent/total lesion area was 10.6 times higher on buccal surfaces in a subject than on lingual surfaces ($P < 0.05$)	The number of new lesions developing or progressing on bracketed buccal surfaces was 4.8 times higher and the caries extent, or integrated fluorescence loss, was 10.6 times higher than in lingual surfaces	High risk
Beyling <i>et al.</i> , Head Face Med 2013 (28)	Clinical	Incidence of demineralization beneath the lingual bracket base at the end of treatment with two bonding protocols	With standard indirect chemical cure bonding, 18 lesions occurred in 120 surfaces (15%). With the addition of an extra layer of a hydrophilic resin, 6 lesions occurred in 120 surfaces (5%), $P = 0.01$	Gender and age were not significantly related to the development of new lesions	High risk

Table 2. Continued

Study	Subject group	Definition of pre-specified main outcome	Summary outcome data	Additional outcomes	Quality assessment*
Deguchi <i>et al.</i> , Angle Orthod 2015 (31)	Clinical	Cephalometric values, degree of root resorption, PAR, OGS, and functional evaluation in lingual and labial treatment	Pre-treatment PAR, labial: 25.0 ± 9.6 , lingual: 28.0 ± 7.2 (ns); post-treatment PAR, labial: 4.0 ± 2.2 , lingual: 4.2 ± 2.2 (ns). Pre-treatment cephalometric measurements (ns). Post-treatment only interincisal angle (T0–T1, labial: 11.2 ± 11.6 ; lingual, 17.9 ± 8.9 , $P < 0.05$). Root resorption, labial: 1.1 ± 0.5 mm, lingual: 1.3 ± 0.6 mm (ns). Jaw movements increased in the same way in both groups	Average treatment time, labial: 29.4 ± 5.6 m, lingual: 32.5 ± 6.7 m (ns)	High risk
Ziebura <i>et al.</i> , Am J Orthod Dentofacial Orthop 2014 (32)	Clinical	Number and positions of lost brackets in the first year of lingual and labial treatment	Bond failures per patient lingual: 2.63 ± 2.77 , buccal: 2.61 ± 3.41 ($P = 0.428$)	Molar brackets failed more than premolar ($P < 0.05$); premolar brackets more than canine and incisor ($P < 0.05$). Tendency for more failures in the mandible (lingual: $P = 0.069$; buccal: $P = 0.036$). Occlusal pads in lingual lowered failures in molars, but not in premolars	High risk
Knösel <i>et al.</i> , Head Face Med 2014 (18)	Clinical	Treatment duration with two types of customized lingual appliances	Treatment duration WIN (Severity1: 17.96 m; S2: 20.49 m); Incognito (S1: 22.7 m; S2: 29.79 m) $P < 0.001$	Significant influence of 'appliance type', and 'case severity' on treatment duration, not dependent on each other; no difference in frequencies of bracket losses	High risk
Demling <i>et al.</i> , Angle Orthod 2010 (26)	Periodontal health	Changes in periodontal and microbial parameters after 1 month of lingual treatment on the lower arch	Mandible lingual side PI: $t0 = 0.3 \pm 0.3$, $t1 = 1.0 \pm 0.7$ ($P = 0.001$); BOP: $t0 = 23.4 \pm 22.5$, $t1 = 46.2 \pm 23.5$ ($P = 0.001$); PPD: $t0 = 2.1 \pm 0.4$, $t1 = 2.2 \pm 0.3$ (ns); prevalence of Aa: $t0 = 25\%$, $t1 = 35\%$; prevalence of Pg: $t0 = 5\%$, $t1 = 5\%$	No difference in control sites in any of the tested variables between $t0$ and $t1$	Unclear risk
Miethke and Brauner, J Orofac Orthop 2007 (27)	Periodontal health	Difference in gingival index (GI), papillary bleeding index (PBI). Plaque index (PI), sulcus probing depth (SPD) between lingual and Invisalign treatment at three consecutive appointments (T1, T2, T3)	GI Invisalign/GI lingual Median T1: $0.68/1.13$ Median T2: $0.56/1.02$ Median T3: $0.34/1.00$ PBI Invisalign/PBI lingual Median T1: $0.21/0.57$ Median T2: $0.19/0.55$ Median T3: $0.11/0.58$ PI Invisalign/PI lingual Median T1: $0.31/0.79$ Median T2: $0.34/0.96$ Median T3: $0.19/0.83$ The SPD of approximately 2.5 mm was similar in both groups in all time points	All the indices improved significantly between the first and third assessments in the Invisalign patients, while they were significantly worse in lingual treatment	Unclear risk

*Low risk, unclear risk, or high risk of bias study; ns, non-significant.

Table 3. Risk of bias summary for included retrospective studies.

	Grauer and Proffit (19)	Pauls (20)	Thalheim and Swestka- Polly (21)	Lossdörfer <i>et al.</i> (29)	Wiechmann <i>et al.</i> (22)	Smith (23)	Geron <i>et al.</i> (24)	Beyling <i>et al.</i> (28)	Deguchi <i>et al.</i> (31)	Ziebur <i>et al.</i> (32)	Knösel <i>et al.</i> (18)
Representative sam- ple of adequate size	Unclear	Unclear	Unclear	High	High	Unclear	High	Unclear	Unclear	Unclear	High
Was there a comparison? What kind?	Low	High	Unclear	Unclear	Unclear	High	High	Low	High	High	High
How participants were allocated to groups?	Unclear	High	High	Unclear	Unclear	High	Unclear	Unclear	Unclear	Unclear	Unclear
Which parts of the study were prospec- tive?	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	High
On what variables was comparabil- ity between groups assessed?	Low	Low	Low	Low	Low	Unclear	High	High	Unclear	High	High
Incomplete outcome reporting	Low	Unclear	Low	Low	Low	Low	Unclear	Low	Unclear	Low	Low
Inclusion criteria ex- plicitly described	Unclear	High	High	High	Unclear	Unclear	High	Low	Low	Unclear	Unclear
Adjusting for con- founders	Low	High	Unclear	Unclear	Unclear	High	High	High	Unclear	High	High
Description of potential biases	Unclear	High	High	High	High	High	High	High	High	Unclear	Unclear
Blinding of the assessor	Unclear	High	Unclear	Unclear	Unclear	Low	High	Low	High	Low	Low
Reporting of drop- outs/excluded cases	Unclear	High	High	High	Unclear	Unclear	Unclear	Unclear	Low	High	Unclear
Reporting of follow- up	Low	Low	Low	Low	Low	Low	Unclear	Low	Low	Low	Low
Overall	Unclear	High	High	High	High	High	High	High	High	High	High

In each criterion, each study was classified as having low, unclear, or high risk of bias. Overall, studies with at least one high risk are considered high risk of bias, studies with at least one unclear risk are considered of unclear risk of bias, while studies with low risk only low risk of bias.

duration was significantly smaller with WIN compared to Incognito system (18). The results of these investigations, however, have to be interpreted taken into account the possibility of the influence of the various biases on the study outcomes.

The third subject group included two prospective CCTs that investigated the effect of lingual orthodontic treatment on various periodontal parameters. The first study tested, in a split-mouth design, the short-term influence of Incognito lingual appliances on various clinical periodontal and microbial indices and showed a worsening of plaque index and bleeding on probing in the bonded sites, while no significant difference was detected on probing depth and periodontopathogenic bacteria (26). The other study tested the effect of Ormco seventh generation lingual treatment as well as Invisalign treatment on clinical periodontal parameters. All the indices apart from probing depth improved significantly between assessments in the Invisalign patients, while they were significantly worse in lingual treatment in all time points (27). Concerning the interpretation of the findings of these two studies, however, it must be taken into consideration that both are single non-randomized studies with unclear risk of bias.

Limitations

There is a shortage of large, high-quality prospective clinical trials investigating clinical outcomes and effects of lingual orthodontic treatment (only one RCT was available). Thus, the majority of the included studies had a retrospective design and the risk of bias was high. An assessment of high risk of bias does not necessarily mean that the study

results are biased; it means that the suspicion of biased results is higher compared to those of a study classified at low risk of bias.

In most cases, no synthesis of studies was feasible since there was only a single study on each subject. In the presence of more than one study on a subject, grouping and/or synthesis of results and conclusions was not feasible due to the clinical heterogeneity of the included studies (e.g. different bracket systems are tested).

Language restrictions might be an additional limitation in the present systematic review.

Conclusions

This systematic review showed encouraging results on the clinical outcome of lingual orthodontic treatment, especially in regards to the achievement of individualized treatment goals and the decreased risk of decalcifications on the bonded surfaces of the teeth. However, additional well-designed prospective clinical trials with large samples are needed to confirm those findings. Several aspects of lingual orthodontic treatment were difficult to be conclusively evaluated due to the study design, the heterogeneity, the small samples, and the high risk of bias seen in the majority of the included studies.

Supplementary material

Supplementary material is available at *European Journal of Orthodontics* online.

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